Journal of Applied Health Sciences and Medicine
ISSN (Online): 2583-1887
www.jpub.org

Volume 2, Issue 2: 41 – 44, 2022

Original Case Study

Pemphigus Vulgaris Confined to the Gingiva

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Received: 12.6.2022 • Accepted: 25.6.2022 • Published: 29.6.2022

Abstract: Pemphigus Vulgaris (PV) is an intraepithelial blistering disease that frequently affects the skin and mucous membranes. The oral mucosa is frequently affected in pemphigus patients. It does not have any gender preferences and can affect both men and women aged 40 to 60. In many patients, oral lesions may be the only initial indication of the disease. Oral lesions may be followed by skin lesions in some patients. PV manifests as blisters and erosions of the skin and the mucous membrane. Timely diagnosis and treatment of oral lesions is important. It is a challenge to diagnose PV with oral lesions because of their nonspecific presentations. The lesions are superficial erosions or ulcerations. Intact bullae are difficult to find but when found may be filled with clear liquid. Lesions may occur anywhere on the oral mucosa including gingiva. Gum inflammation or desquamative gingivitis is less common compared to other muco - cutaneous conditions such as mucous membrane pemphigoid (MMP) or oral lichen planus. This paper describes a case of a patient presenting with a three years history of painful and burning sensation of gingiva, which was earlier misdiagnosed and treated as gingivitis. Finally, it was diagnosed as a case of pemphigus vulgaris.

Keywords: Pemphigus Vulgaris, Woman, Gingiva.

1. Introduction

PV is an autoimmune disease characterized by intraepithelial blistering of the skin and mucous membranes [1,2]. PV is distinguished by acantholysis of the epithelium [3]. It affects nearly equal numbers of males and females and is more prevalent in middle-aged and elderly patients [4]. Significant improvements are associated with systemic corticosteroid therapy [2]. Nonetheless, medical therapy complications remain a cause for concern. Patients with PV frequently experience oral lesions, which are the most prevalent initial symptom of PV [2]. The gingiva is the least frequently affected site, and desquamative gingivitis (DG) is a common symptom of the disease. The buccal mucosa is the most frequently affected site, followed by the palatal, lingual, and labial mucosae [5]. In many PV patients, the development of skin lesions comes after the development of oral lesions [3]. Because of this, if oral PV is discovered early on, treatment can start to stop the disease from developing into skin involvement [3]. However, early oral lesions of PV are

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frequently thought to be difficult to diagnose because they can initially present as superficial erosions or ulcerations and infrequently form intact bullae [5].

2. Case Report

We present a case of 30 year old woman who presented to our clinic with symptoms of mild pain and burning sensation of the mouth while brushing. She had these symptoms for three years. The pain increased with hot and spicy foods and did not improve despite several visits to the local hospital. She was treated as gingivitis. She revealed no systemic problems. She was an occasional drinker. Her vitals were all within normal range.

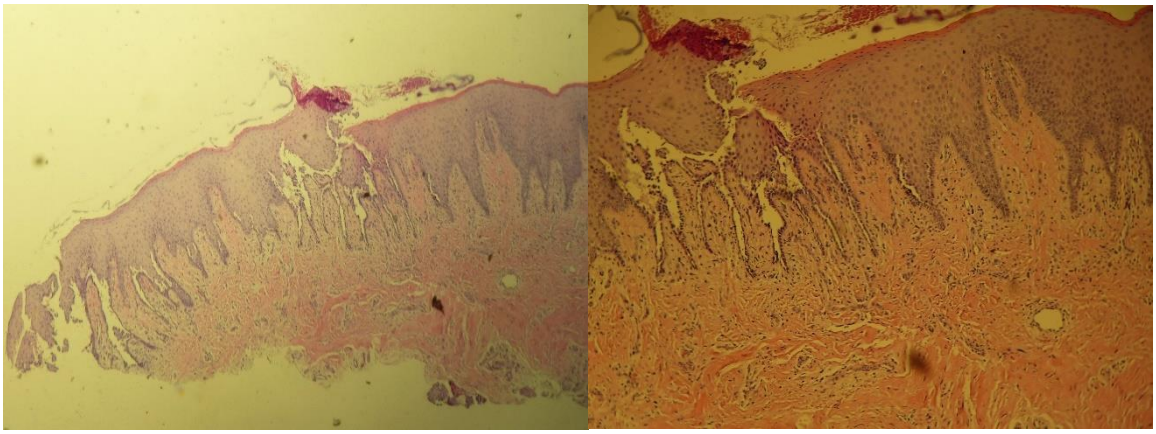
On extra oral examinations, there was facial symmetry, no swellings or lumps, no skin lesions. Both the Temporo-mandibular joints were normal.

On Intra-oral examinations, there was mild erythema of the gingiva in the anterior mouth only involving both the attached and marginal gingiva. There was mild desquamation of the gingiva with occasional white areas/patches at few points on the gingiva. There was no involvement of any other area of the mouth. The lips, tongue, buccal mucosa, palate and other structures were normal. The gingiva as seen in the Figure below 1 (a), (b) and (c):

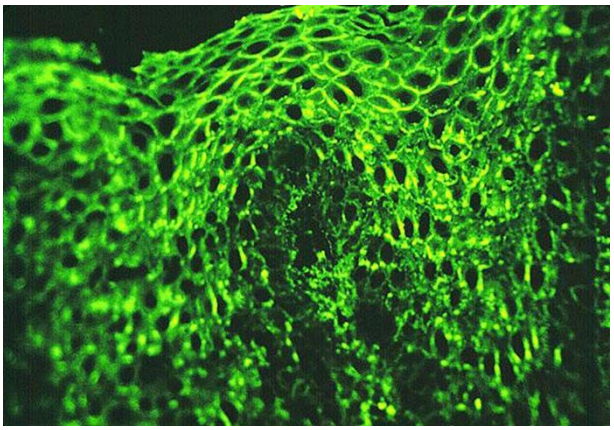
1(a)

1(b) 1(c)

An incisional biopsy was performed and specimens sent for histopathological examinations (HPE) and direct immune-fluorescent study (DIF). Both the reports were consistent with pemphigus vulgaris. HPE and DIF pictures as seen in the Figure 2 (a), (b) and (c) below:



2 (a) Low power field (HPE) 2(b) High power field (HPE)



2 (c) DIF finding-Intra-epithelial deposition of immunoglobulin

The patient was treated with topical corticosteroids i.e. dexamethasone mouth wash and topical application of Triamcinolone Acetonide 0.1% in orabase. Chlorhexidine mouthwash was given to clean the teeth. A systemic steroid was not used because there was no skin lesions neither involvement of others mucosa. The patient was followed up at 2 weeks, 4 weeks and monthly there-after until the oral lesions healed completely. The pictures as in Figure 3 (a) and (b) shows before and after treatment images:

3(a)-Before Treatment 3(b) - After treatment

3. Discussion

Early diagnosis and treatment of PV is important since it is a fatal disease [3]. The mainstay of treatment is corticosteroid in high dose(1). Usually systemic corticosteroid 1-1.5mg/kg/day is given in a single or divided dose [1,2]. Once the remission occurs the drug can be tapered down and given as maintenance dose [3]. Adjuvant drugs are often given to reduce the side effects of steroid [6]. Adjuvant drugs used are Azathioprine, Cyclosporine, Cyclophosphamide, Mycophenolate mofetil, Methotrexate [7], Tetracycline and Dapsone [6,8]. The choice of drugs to be given depends on the severity of the disease and the clinicians' judgement [5,6]. Histopathology with Immune-fluorescent study is the gold standard for diagnosis of pemphigus [1,9]. Therefore, the following requirements must be met for a definitive diagnosis of PV: confirmation of acantholysis in biopsy specimens, and confirmation of auto antibodies in tissue, serum, or both PV was confirmed by the presence of Nikolsky's phenomenon, acantholysis in biopsies, and the DIF test for detecting antibody deposition between epithelial cells.

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